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## Case Report

# Thumb Polydactyly with a Floating Ulnar Thumb

Kenjiro Hasegawa\*, Yuzaburo Namba, and Yoshihiro Kimata

*Department of Plastic and Reconstructive Surgery, Okayama University Graduate School of Medicine,  
Dentistry and Pharmaceutical Sciences, Okayama 700-8558, Japan*

Thumb polydactyly is reported to be the most common congenital anomaly of the hand in Japan. The floating type is not particularly rare, accounting for 0.9 to 15% of all cases of thumb polydactyly. However, to the best of our knowledge, there has been only one case of thumb polydactyly with a floating ulnar thumb, reported by Onizuka. Herein, we report a case very similar to that reported by Onizuka. In our case, the vessels feeding the floating ulnar thumb branched from the superficial palmar arterial arch, and X-rays revealed triphalangism. In surgery, we not only reconstructed the morphology of the thumb, but also tried to preserve the sensation in the reconstructed thumb by transposing the digital nerve of the floating ulnar thumb to the radial thumb. In addition to thumb polydactyly, our case also showed hypoplasia of the thenar muscles.

**Key words:** polydactyly, floating ulnar thumb, thumb hypoplasia

A considerable number of cases of thumb polydactyly with a floating radial thumb have been reported [1–4]. We encountered a case of thumb polydactyly with a floating ulnar thumb and hypoplasia of the thenar muscles, and compared the findings in our case with those of previously reported case [5].

## Case

The patient was a 2-year-old boy with bilateral thumb polydactyly. He was born after 34 weeks of gestation as the third baby of triplets, with a birth weight of 1794 g. He also had tricuspid regurgitation. The other 2 baby boys who were born at the same time had no hand anomalies or complications.

His left thumb polydactyly was classified as type 4 triphalangism (Fig. 1), according to the Japanese

Society for Surgery of the Hand: Modified International Federation of Societies for Surgery of the Hand classification (Modified IFSSH Classification) [6], while his right thumb polydactyly with a floating ulnar thumb was classified as type 7 (floating type). The stem of the right-hand floating ulnar thumb was located on the radial side of the distal metacarpal of the index finger, and not the thumb, and X-rays revealed triphalangism. The radial thumb had no nail, and X-ray images showed the proximal phalanx and metacarpal, but not the distal phalanx (Fig. 2). Hypoplasia of the thenar muscles on the right hand was also observed. The thumb hypoplasia was classified as type I according to the Blauth classification (Fig. 3) [7].

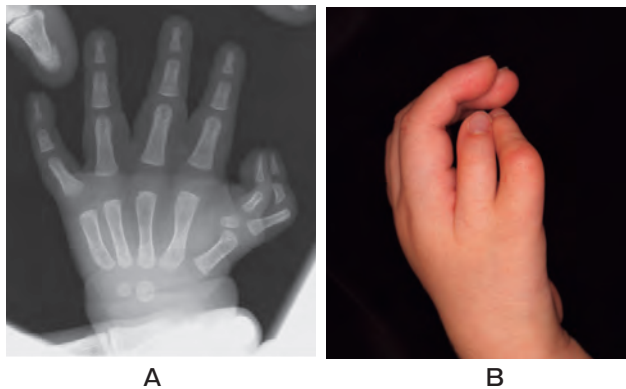
At 1 year of age, the patient underwent surgery for tricuspid regurgitation, and surgery for right thumb polydactyly was performed after improvement of his cardiac function.

In the first operation, the radial thumb without the nail was excised at the distal level of the proximal phalanx, and the ulnar thumb was transplanted as a

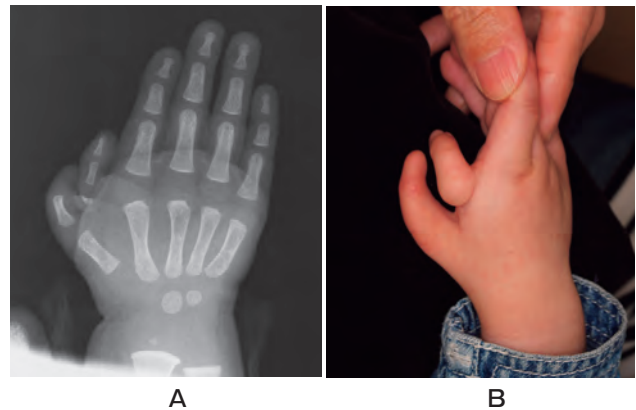
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\*Corresponding author. Phone: +81-86-223-7214; Fax: +81-86-235-7210

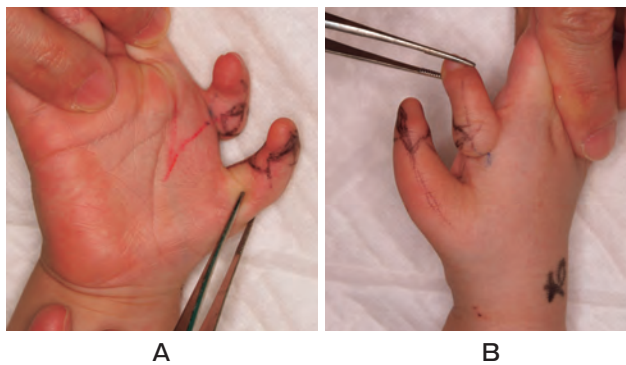
E-mail: k-hase@md.okayama-u.ac.jp (K. Hasegawa)



**Fig. 1** The left thumbs of the 2-year-old boy with bilateral thumb polydactyly. The anomaly was classified as type 4 triphalangism, according to the Modified IFSSH Classification, type VII according to Wassel's classification, and type VII A according to Wood's classification. **A**, X-ray findings; **B**, polydactyly of the left thumb.

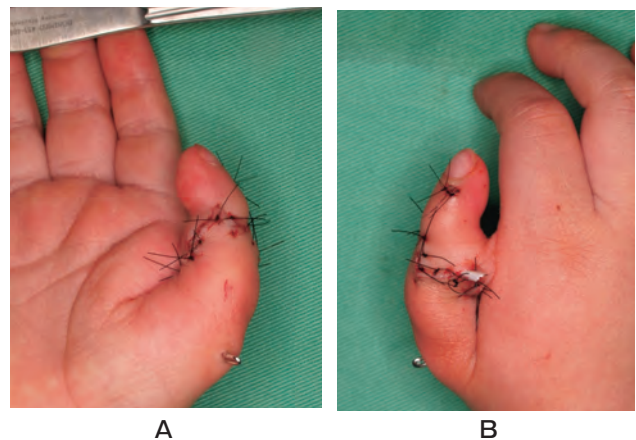


**Fig. 2** The right thumbs of the 2-year-old boy with bilateral thumb polydactyly. The anomaly was classified as type 7 (floating type) according to the Modified IFSSH Classification, type VII according to Wassel's classification, and type VII A according to Wood's classification. **A**, X-ray findings; **B**, polydactyly of the right thumb.



**Fig. 3** Preoperative photographs of the right hand. **A**, Hypoplasia of the thenar muscles was seen; **B**, The nail of the radial thumb was missing.

pedicle flap (Fig. 4). The flexor and extensor pollicis longus tendons were missing; therefore, tendon suture was not performed. Before the second operation, multidetector-row computed tomography (MDCT) angiography showed that the feeding vessels of the floating ulnar thumb branched from the radial digital artery of the index finger, originating from the superficial palmar arterial arch (Fig. 5A). In the second operation performed 4 weeks later, the neurovascular bundle of the ulnar thumb was transposed to the radial thumb and the pedicle flap was excised (Fig. 6). The postoperative course was uneventful, and 2 months after the surgery, the patient was able to pinch and grip with the reconstructed thumb (Fig. 7, 8). Seven months after the surgery, the patient was able to stably grasp a pencil. X-ray findings revealed no



**Fig. 4** Photographs taken immediately after the first operation on the right hand. **A**, **B**: The radial thumb without the nail was excised at the distal level of the proximal phalanx, and the ulnar thumb was transplanted as a pedicle flap.

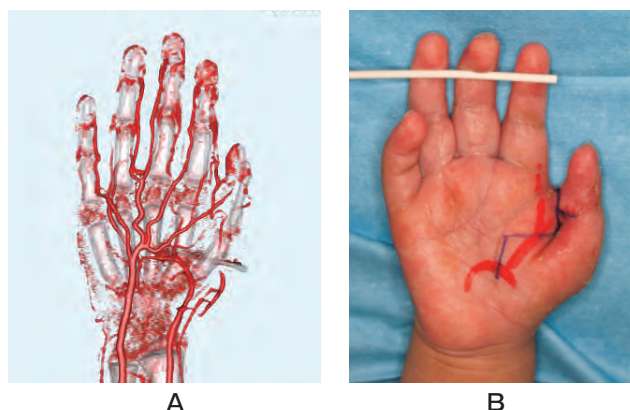
ossification at the site of osteosynthesis at the level of the proximal phalanx; however, clinically no instability was recognized in this region. In the event of the appearance of instability in the future, bone grafting will be performed (Fig. 9).

## Discussion

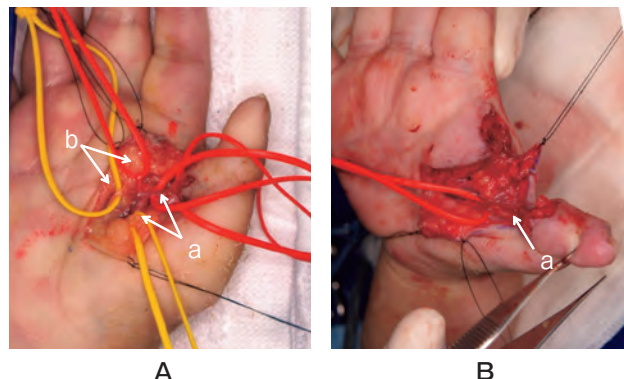
Graham *et al.* reported the relationship between thumb polydactyly and thumb hypoplasia in 4 pedigrees in which thumb polydactyly and thumb hypopla-

sia were found [8]. Gabel *et al.* reported a case of hypoplasia of the left thumb and polydactyly of the right thumb, and pointed out the possible relationship between thumb polydactyly and thumb hypoplasia [9]. Cases of thumb polydactyly and thumb hypoplasia on the same side, as in the present case, have been reported in some patients, including 2 patients reported by Graham *et al.* [8], 3 patients by Miura *et al.* [10] and 9 patients by Kita *et al.* [4]. However, no cases of floating-type thumb polydactyly with thumb hypoplasia have been reported previously, to the best of our knowledge.

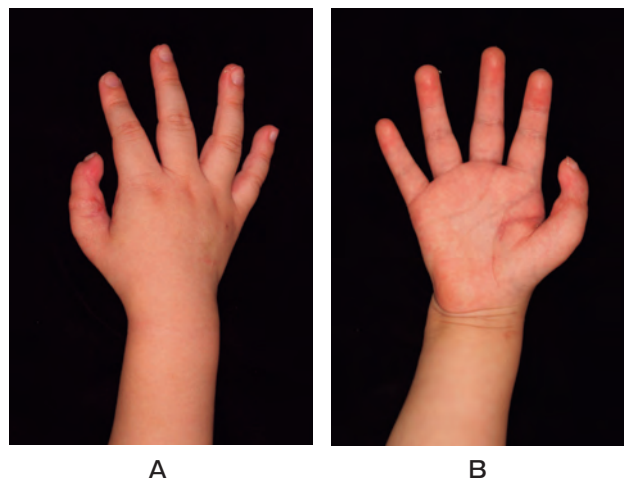
Flatt described that thumb polydactyly was the third most commonly encountered congenital anomaly of the hand after syndactyly and camptodactyly [11]; however, thumb polydactyly has been reported to be



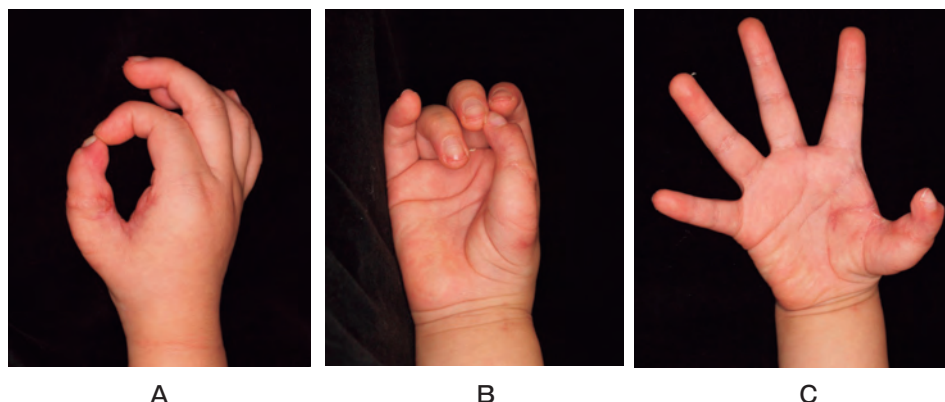
**Fig. 5** Photographs taken before the second operation on the right hand MDCT angiography showed that the feeding vessels of the floating ulnar thumb branched from the superficial palmar arterial arch. **A**, MDCT angiography; **B**, a photograph taken just before the pedicle flap amputation.



**Fig. 6** Photographs taken during the second operation. **A**, The neurovascular bundle of the floating thumb (a) and the radial digital nerve and artery of the index finger (b) were identified (red tapes: arteries, yellow tapes: nerves); **B**, The radial digital artery of the index finger was cut at its distal side and the neurovascular bundle of the floating thumb was transposed to the radial thumb.



**Fig. 7** Photographs taken 2 months after the operation. **A**, Dorsal view of the right hand; **B**, Palmar view of the right hand.



**Fig. 8** Movements of the thumb 2 months after the operation. **A**, tip pinch; **B**, chuck pinch; **C**, abduction.



the most common congenital anomaly of the hand in Japan [12, 13]. Wassel's classification [14], which is based on the level of bifurcation on plain radiographs, is widely used to classify thumb polydactyly. In Japan, the Modified IFSSH Classification [6], a modified version of Wassel's classification, is often used. One of the main differences between the 2 classifications is the non-inclusion of the floating type in Wassel's classification.

The present patient with a floating ulnar thumb on the right hand showed triphalangism, based on which the anomaly was classified as type VII according to Wassel's classification. Furthermore, it was classified as type VII A according to Wood's classification

[15], or thumb polydactyly with a triphalangeal thumb. The present case was also classified as type 7 (floating type) according to the Modified IFSSH Classification.

According to reports analyzing cases that are not included in Wassel's classification, the floating type of thumb anomaly is not rare, accounting for 0.9 to 15% of all thumb polydactyly cases [1-4]. However, to the best of our knowledge, there has been only 1 previously reported case of thumb polydactyly with a floating ulnar thumb, by Onizuka [5].

In Onizuka's report, only 4 photographs were included, and other details of the patient such as the age, sex, and presence/absence of thumb hypoplasia remain unknown. However, the case was classified as

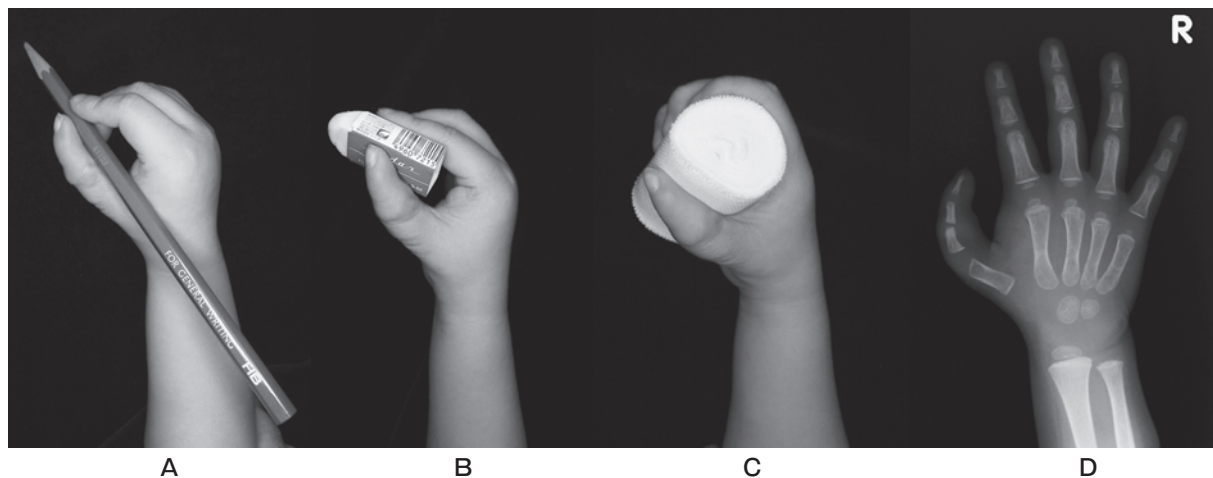


Fig. 9 Movements of the thumb and X-ray findings 7 months after the operation. A, pulp pinch; B, side pinch; C, power zgrip; D, postoperative radiograph.

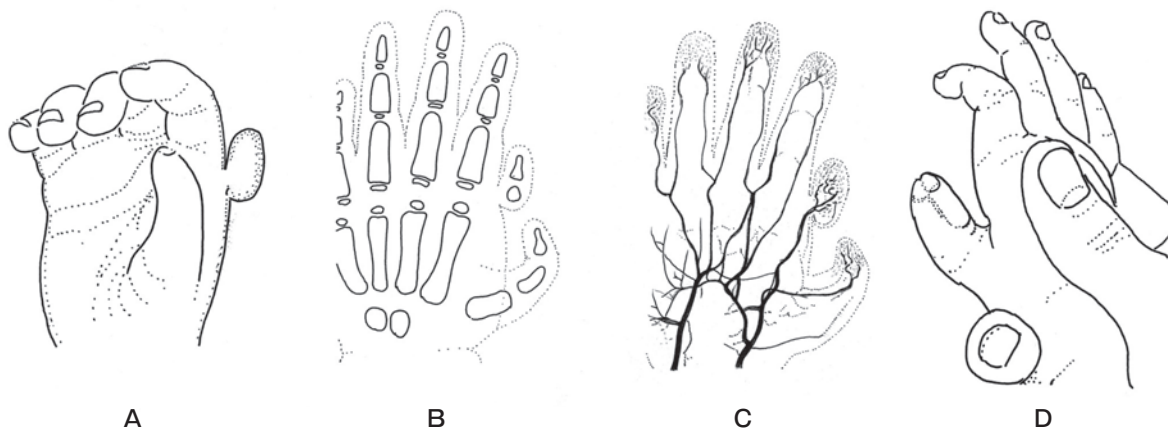


Fig. 10 The case reported by Onizuka [5]. A, before operation; B, preoperative radiograph; C, arteriogram; D, 6 months after the operation.

type IV according to Wassel's classification and type 7 according to the Modified IFSSH Classification, based on the X-ray findings. Angiographic findings of the hand showed that the digital artery to the floating ulnar thumb originated from the radial artery. A postoperative photograph indicated that the Bilhaut-Cloquet procedure [16] was used for surgery to adjust to the thickness of the unaffected finger (Fig. 10).

In the present case, the right-hand radial thumb had no nail and had a tapered shape. Therefore, the tip of the radial thumb was excised and the floating ulnar thumb was transplanted to the distal part of the radial thumb to reconstruct the thumb with a nail. Although it was considered possible to perform a single-stage operation using an island flap, the operation was performed as a two-stage procedure due to concerns about congestion. The ulnar thumb was transplanted as a pedicle flap in the first operation. In the second operation, when the pedicle flap was excised, the neurovascular bundle of the ulnar thumb was transposed to the radial thumb to try to preserve the sensation of the reconstructed thumb. It was considered important to preserve the sensation at the tip of the reconstructed thumb so that the patient could perform pinching and gripping motions after the operation.

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